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Parents' Coping with Children's Newly Diagnosed Long-QT Syndrome: A Qualitative Study of the Function and Utilization of an Online User Group

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PARENTS' COPING WITH CHILDREN'S NEWLY DIAGNOSED LONG-QT SYNDROME: A QUALITATIVE STUDY OF THE FUNCTION AND UTILIZATION OF AN ONLINE USER GROUP

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Dissertation Approval

This is to certify that the thesis presented to us by Allison Burns-Pentecost on the 31st day of October, 2012, in partial fulfillment of the requirements for the degree of Doctor of Psychology, has been examined and is acceptable in both scholarship and literary quality.

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Dedication

This research is dedicated to all families who struggle with a child’s medical condition.

Abstract

Long QT syndrome is a sudden death syndrome that occurs in about 1 in 2,000 births. LQTS is caused by genetic mutations that affect the electrophysiology of the heart, resulting in a prolonged QT interval and possible cardiac arrhythmia, syncope or sudden death. Treatments include medications, implantable cardioverter defibrillators and activity restrictions. Little research exists regarding the psychosocial factors of the illness or the ways in which families cope with diagnosis and management of the chronic illness. The current study used archival data from an online user group. Participants were members of the group who discussed concerns related to LQTS. Qualitative methods were used to investigate the function and utilization of the online user group. Nine participants were followed over four years as they coped with the psychosocial challenges of LQTS. This study was the first to examine, specifically, the function and utilization trends of the online user group. Results found that functions included seeking connection and normalization, seeking emotional support and seeking information regarding parenting children of LQTS. Utilization trends changed over time and demonstrated role reversals from seeking to providing behaviors. The grounded method generated a theory that LQTS creates additional psycho-social-emotional demands on parents. When these demands exceed the resources of the current social-cultural milieu, additional resources such as online user groups are needed. Results further suggest that utilization of an online user group for LQTS may support healthy emotional coping through peer support from those with similar experiences. Further research evaluating additional resources for meeting the needs of families coping with a new diagnosis of LQTS is warranted.

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CHAPTER 1

Introduction

Statement of the Problem

Long-QT syndrome (LQTS) occurs in 1 in 2000 births, five times more commonly than previously reported (Schwartz et al., 2009). The highest prior estimate was 1 in 10,000 births (Moss & Robinson, 2002). LQTS, a cluster of congenital, cardiac disorders characterized by prolongation of the QT interval, is caused by mutations in the genes that control ventricular repolarization (Giuffre, Gupta, Crawford & Leung, 2008). LQTS is a sudden death syndrome in which affected persons may live asymptomatic for years and then die suddenly from arrhythmias or sudden cardiac arrest. Factors that may lead to fatality include exercise and exertion, loud noise with startle, and inactivity such as sleep. Because of this, families of children with LQTS are often advised to avoid mental stress, strenuous sports and loud noises. Side effects of beta-blockers, a common treatment for LQTS, have included depression, fatigue and mood changes (Giuffre, Gupta, Crawford & Leung, 2008). Given the commonality of life threatening risk factors, patients and families of LQTS may experience psychosocial stress related to being diagnosed with and living with LQTS.

Furthermore, recent research has found that positive emotions such as happiness may have a protective effect against cardiac events in LQTS and that stressful events may increase the chance of a cardiac event, pointing to the importance of managing stress and emotions for persons with LQTS to prevent life threatening events (Lane, Reis, Peterson, Zareba, & Arthur, 2009). Given the integral role of avoiding triggers, managing stress and maintaining a positive mood for long term health in patients with LQTS, it is

surprising that so few studies have examined the psychosocial aspects of LQTS (Anderson, Oyen, Bjorvaton & Gjengedal, 2008; Giuffre, Gupta, Crawford & Leung, 2008; Hendriks et al., 2005; Lane, Reis, Peterson, Zareba, & Arthur, 2009).

Much of the existing research regarding psychosocial aspects of chronic illness in children and their families has focused on other chronic health illness such as spina bifida, cancer, asthma and arthritis (Barlow & Ellard, 2006). Even within this broader area of chronic health, findings regarding the interaction of chronic health and psychosocial factors have been mixed. Some researchers have found increased anxiety, depression, behavioral and adjustment problems in children with childhood cancer (Houtzager, Grootenhuis & Last, 1999); however, others (Bennett, 1994) have found no differences on measures of depression or self-concept in children with a variety of chronic health conditions. Studies of psychosocial factors across various chronic health conditions (cancer, diabetes, arthritis) have produced various findings, suggesting that research from one chronic health condition may not generalize to others (Barlow & Ellard, 2006; Taylor Fuggle & Charman, 2001).

Research exploring chronic health conditions has found that social support primarily that of the family is a protective factor (Dobbie & Mellor, 2008). There is a direct relationship between the patient's ability to cope with his or her illness and the family's ability to cope, suggesting a cyclical relationship between the family's health and the health of the patient with chronic illness (Dobbie & Mellor, 2008). Methods for seeking social support have extended beyond traditional means, such as the medical community, family and friends. Families of children with chronic illness may now use internet -based groups, including those exclusive to specific disorders such as LQTS.

Little is known about the use of these internet-based groups in the role of seeking social support and of coping in families dealing with LQTS.

Given the mounting evidence that the prevalence of LQTS is higher than previously indicated (Schwartz, et al., 2009), the importance of emotional regulation in managing LQTS (Lane, Reis, Peterson, Zareba, & Arthur, 2009), and the lack of existing research examining psychosocial factors of chronic illness, specifically LQTS, it is apparent that further research is needed to understand how individuals with LQTS experience psychosocial factors and the methods they use to cope and seek support. Investigating the role of an internet-based user group may provide further insight into the important dynamic between the role of chronic health conditions, the role of the family, and the use of the internet as a method of coping, seeking support and ultimately maintaining health.

Background

Long QT syndrome (LQTS) is characterized by a prolonged QT interval on the electrocardiogram, secondary to mutations in ion channels in the heart. This results in an electrical instability in the heart and may lead to a rapid heartbeat or ventricular arrhythmia, known as “de pointes” or twisting of the points; this may further result in a loss of consciousness, or syncope, and a sudden cardiac arrest leading to sudden cardiac death. When the heart beats too fast, it is not effective in maintaining the circulation and the blood does not flow to the brain, leading to a loss of consciousness, often without warning. This abnormality occurs due to the ion channels of the heart’s electrical system during ventricular depolarization or during repolarization. LQTS is caused by an inherited autosomal dominant or by autosomal recessive genetic mutation or due to a

sporadic mutation in the individual affected. LQTS may also be acquired, due to the effects of certain drugs on ion channels, often in the setting of sinus nucleotide polymorphisms, or minor changes in ion channel genes related to LQTS. (QTsyndrome.ch, 2007).

LQTS is considered to be a leading cause of sudden death in young people (Schwartz et al., 2009). The prevalence rate of LQTS is currently unconfirmed and is thought to be underestimated because it often goes undetected until syncope or a sudden cardiac arrest occurs. It is thought that perhaps 10% to 12% of Sudden Infant Death (SIDS) cases may actually be a result of undiagnosed LQTS (Baruteau et al., 2009). A recent study provides the first data-based estimate of the prevalence rates to be approximately 1:2000 in Caucasians (Schwartz et al., 2009). These estimates are much higher than previous reports of 1:5000 (Goldenburg et al., 2008). Fugate et al. (2010) reported that the clinical course, risk factors and benefits from treatments are similar in African-Americans, as compared with Caucasians. The most recent prevalence data, however, excluded non-whites and therefore the prevalence rate are not known in these groups (Schwartz et al., 2009).

Since the first published case of LQTS in 1957 by Jervell and Lange-Nielsen, additional variants have continued to be discovered; currently more than 700 mutations have been located across 13 LQTS genes (Moss, 2003). Ninety-five percent of identified mutations occur across the first three types of LQTS with LQTS 1 at 43%, LQTS 2 at 45% and LQTS 3 at 7% of individuals affected. These various genotypes of LQTS are largely related to clinical symptoms or phenotype, the physical expression of the gene.

Clinical symptoms and risk of cardiac events vary among the different genotypes of LQTS. Persons affected by LQTS 1 tend to experience the greatest number of their events during exercise, sports or physical exertion, whereas persons with LQTS 2 have the greatest number of events coupled with emotional arousal and LQTS 3 triggers cardiac events during rest or sleep (Moss, 2003). Cardiac events are significantly more frequent in LQTS 1 and LQTS 2; LQTS 3 tends to produce fewer cardiac events but those that occur are more likely to be fatal. Cardiac events are also significantly higher during adolescence among all three types (Moss, 2003). Given the commonality of triggers for major cardiac events, management of the environment and psychological health is particularly important. Furthermore, when a medical condition such as LQTS occurs primarily during childhood and adolescence, the entire family is needed in order to cope effectively.

Clinical symptoms and triggers for arrhythmias, syncope and sudden death vary across the identified types of LQTS, as described previously. They also vary within the same genotypes found within families who have the same genetic expression of LQTS. For example, one family member may be asymptomatic for years and then another family member with the same genetic expression of LQTS may have persistent and recurrent syncope or even experience early sudden death. This variation of phenotype expression within the same genotype variant of LQTS leads researchers to conclude that environmental factors and state traits also play a role in clinical symptoms of the syndrome (Moss, 2003). The interaction of family, environment and emotion is intimately involved in the psychosocial management of LQTS. This sudden death syndrome requires the services of the clinical health psychologist in collaboration with

pediatric cardiologists as well as general medical practitioners, in the medical home, to research and provide the best management and treatments for this chronic medical condition.

Literature Review Summary

In sum, the review of current research includes mechanisms for learning and factors for applying various coping mechanisms, including variations among every day stressors, traumatic life events and chronic medical conditions. Review of the research on coping with chronic medical conditions, in general, has various findings. Research conducted in the late 1960s reported evidence of maladaptive psychological functioning among persons with childhood chronic illness (Pless & Roghmann, 1971; Cadman et al., 1987 as found in Thompson & Raezer, 1998), whereas more recent findings have concluded that the impact of childhood chronic medical conditions can be significant; however, the majority of children and their families respond adaptively and learn to adjust and cope well (Thompson & Raezer, 1998). Barlow and Ellard's (2006) review of meta-analyses suggested that parents and siblings may also present with psychosocial needs related to the presence of a chronic medical condition in their family member.

Research regarding the psychosocial factors related to coping with LQTS is miniscule in comparison with that available regarding other chronic medical conditions of childhood. Research suggests those coping with LQTS are frustrated with the medical community (Farnsworth et al., 2006); they would benefit from regular and sequential information regarding LQTS (Anderson et al., 2008; Hendriks et al., 2005) and in general are less concerned with their own health status and more concerned with their children's health prognosis (Kroode et al., 2000), during genetic testing. Research suggests that

older children and adolescents may have greater difficulty responding to a new diagnosis of LQTS than younger children (Farnsworth et al., 2006). Also, studies examining those newly diagnosed found that people often express feelings of relief and of validation for the symptoms experienced (Anderson et al., 2008). Giuffre et al. (2008) suggested the fear and uncertainty associated with LQTS may internalize for some children with LQTS. Lane et al., (2009) found that positive emotions and happiness in particular may buffer individuals from future cardiac events associated with LQTS.

Research regarding the psychosocial factors related to LQTS is limited and warrants further investigation. LQTS is different from other more commonly studied childhood chronic illness. The variation of symptoms of LQTS has a considerable range, from asymptomatic to sudden death. In addition, an estimated 10-15% of SIDS cases are attributable to undiagnosed LQTS (Tester & Ackerman, 2007). Based on the previous review, health care providers and the general community need more information and better education regarding the signs and symptoms of LQTS. Additionally, the fields of pediatric cardiology and clinical health psychology may better assist patients through further insight and understanding of the psychosocial factors related to living with LQTS.

Those coping with chronic medical conditions, such as LQTS may often utilize the internet as they cope, particularly when newly diagnosed (Forkner-Dunn, 2003). Participants of an online community may be more likely to contribute personal information, artifacts or emotional content because the environment feels sheltered and anonymous. Research also suggests that the internet creates an environment that transcends gender norms (Bellman et al., 1993; Mo, Malik and Coulson, 2009; White & Dorman, 2000) and some racial biases (Anderson, 2003). These findings suggest that

computer mediated communication (CMC) may reduce some prejudice based on visual, racial and contextual cues, allowing for increased equality and status in computer mediated working relationships.

Research in this field of internet-based interventions is burgeoning. A myriad of formats exist for internet-based user groups; these vary among mediated vs. non-mediated, formal vs. informal and structured vs. unstructured. Also, the types of communication tools and site designs vary among user groups, including access to synchronous vs. asynchronous posting options. A review of the research examining the outcomes for internet-based interventions and support groups varies. Reports offer some benefits for CMC (Ambrosino & Jaser, 2010; Johnson, Ravert & Everton, 2010; Kalichman et al., 2003) and also some drawbacks (Lieberman & Goldstein, 2006; McCormack, 2010; Eysenbach et al., 2004; Saltzer et al., 2010). To date, no study has specifically examined an internet-based user group to measure the function and utilization patterns of those coping with LQTS. In fact, Eysenback (2004) suggests that unstructured support groups that do not have formal intervention components are particularly understudied because they are typically unfunded. The current study seeks to provide further understanding of the function and utilization of an informal, mediated, unstructured internet-based group, dedicated to those coping with LQTS. A comprehensive review of the literature may be found in Appendix A.

Purpose of the Current Study

The current study utilized qualitative methods to examine the function and utilization patterns of a LQTS internet-based user group, including the characteristics of communication that the participants displayed. Specifically, this study followed 9

members of a LQTS user group and examined participants' characteristics as related to the assumed purpose of use after receiving a diagnosis of LQTS. Understanding the utilization patterns of this online user group and the characteristics of the participants during the first several years post diagnosis of LQTS offers additional information about the factors that participants experience, as well as the mechanisms for coping with those factors.

Previous qualitative studies have examined this LQTS internet-based user group for content themes such as anxiety, compliance, parental control and grief and loss (Haynes-Weller, 2011; Janney, 2011; Rovinsky, 2010; Steinhauser, 2010). Recent research is just beginning to uncover the necessity of information and support for people diagnosed with LQTS (Haynes-Weller, 2011; Janney, 2011; Rovinsky, 2010; Steinhauser, 2010); however, more research is needed.

Understanding the utilization patterns of this online user group and the characteristics of the participants with a new diagnosis of LQTS provides additional information about the factors that participants experience and the mechanisms for coping with those factors. Because psychosocial factors are relevant to the maintenance of health in persons with LQTS (Lane, Reis, Peterson, Zareba, & Arthur, 2009) and to the coping of the family unit (Dobbie & Mellor, 2008), more information regarding the methods for coping with the stressors of diagnosis is necessary for the patients, the families, the medical community and the mental health professionals affected by LQTS. Given the prevalence rates of LQTS and the lack of current information regarding internet user groups related to LQTS, increasing awareness regarding these issues is paramount as rising numbers of families and medical professionals are affected.

The results of this study contribute to the understanding of the psychosocial factors for managing LQTS, the interrelationship of biopsychosocial mechanisms of health and behavior, and to the professions of clinical health psychology and pediatric cardiology. Findings from this study, in combination with other related works will help to inform medical and mental health providers as well as families of children with LQTS regarding the utility of online user groups and also the broader psychosocial factors related to LQTS. Advancing the empirical understanding in these areas has far-reaching implications for both medical and mental health care, and the role of mind-body connections in maintaining health.

Research Question

What is the utilization and function of an LQTS online user group for members after a new diagnosis of LQTS?

Research Question Terms Defined:

Function*:* The suggested purpose that the online group serves for the participants. Purposes for posting may include but are not limited to: information sharing, information gathering, processing a recent event, seeking support or normalization.

Utilization: The frequency of posting, duration and patterns of use (including participant communication characteristics and style) of the online group.

CHAPTER 2

Method

Overview

The current study investigated an archival data set and utilized qualitative methods to explore participant characteristics, utilization and function of an online user group post-diagnosis of LQTS. Specifically, the study investigated how members interacted with an informal, moderated, online computer group. The study also explored the characteristics of those members with the goal of generating theory. Members communicated by posting discussion topics online and responding to posts within various discussion threads. The user group included in this investigation is informal, meaning that the frequency, method and purpose for interaction are at the volition of the user. The group is moderated; moderators are also active participants in the group and in addition to monitoring the group activity, they also post comments and provide information to the group participants. Although the group is considered part of the public domain, it is closed and requires registration in order to participate.

Research Design

The grounded theory method of qualitative analysis was used to describe and interpret emerging patterns and concepts from the raw data that consisted of transcripts collected from the discussions of the user group posts. The applied use of the grounded theory paradigm included a systematic procedure to build theory and generate a depth of understanding relative to the concepts and relationships that emerged in the data (Glaser, 1998). These processes produced information about how the participants interacted with

the informal online user group and allowed for the discovery of other broader social-cultural theories regarding the participants' experiences that may not be derived from statistical analysis alone (Strauss & Corbin, 1998). In addition to identifying emerging themes through the use of grounded theory, a conversational analysis (Mann & Fiona, 2009) was used to gain greater insight and understanding relative to the participants' computer-mediated interpersonal interactions.

Design Justification. A qualitative research method was needed for this study because the relevant variables under investigation have yet to be identified. The application of the systematic methods of grounded theory and conversational analysis engendered concepts and relationships with the aim to develop theory, grounded in the contextual and individual experiences garnered from the internet-based conversational data analysis. The transactional system of grounded theory is a method of analysis that allowed the interactive nature of the user group communication to be examined. Specifically, the relationship between participant characteristics and user- group utilization was investigated. Developed by Glaser and Strauss in the 1960s, grounded theory employs a systematic, procedural examination that meets "good" scientific criteria; procedures for validity, reliability and standardization will be included, as further detailed in the procedure and data analysis sections (Marshall and Rossman, 2011). Qualitative methodology is of value because the research questions are exploratory and descriptive in nature.

Internet Based Research. This research is internet-based; the context and setting of the internet cannot be removed from this investigation and therefore a qualitative research design was required to seek understanding of the utilization and function of

online user groups for the studied population. The population of internet users is not representative of the general population because not everyone has access to the internet. Qualitative studies, however, do not require representative samples (Eysenback & Wyatt, 2002) and therefore was further appropriate for this study design. The purpose of this study is not to generalize the results to the general public but rather to systematically study the social environment of an online group for the LQTS community with the aim of clarifying concepts and generating hypotheses for future study.

Internet-based research transcends global boundaries and affords anonymity to the comfort of the user. Human behavior is substantially influenced by the context in which it occurs; therefore, qualitative methods allow these phenomena to be investigated in the natural internet environmental context in which they occur. Increasing an understating of the function and utilization of online internet-based user groups for LQTS is important in developing effective psychosocial supports within the medical and psychological community for these groups.

Selection Procedure

Participants. Twelve threads in the topic area of 'new diagnosis' were identified; 10 of these contained new diagnosis stories. One participant was excluded for not having LQTS himself or herself or in his or her family. In addition to the collection of these 9 participants' initial, introductory 'new diagnosis' posts, all of their subsequent posts, including threads authored and reply posts contributed to peer authored posts were collected and included in the analysis. No recruitment was necessary; the sample was selected from an existing population of online user group participants who are members

of a list-serve that discuss topics related to LQTS. Criteria for membership in the LQTS list-serve include being over the age of 18 and having an expressed interest in LQTS.

Nine participants were selected from the online user group of about 955 international members for inclusion in this study. Participants who posted a new diagnosis story within the previous 4 years or between April 2008 and October 2011 were considered for inclusion in the study. After these participants were identified, all of their remarks posted to the online user group were collected. Dates of these posts ranged from April 2008 to April 2012. Data were collected and coded between November 2011 and May 2012. A total of 198 posts were included in final analysis.

***Inclusion criteria*:** All participants were members of the LQTS user group who posted to a discussion in any topic area related to a "new diagnosis," between April 2008 and October 2011. A chaining sample was used; therefore, any participants who communicated within these topic areas were collected for analysis. In addition to the 9 identified index participants, posts from moderators and peers were also collected for analysis.

Exclusion criteria: User group participants who do not have LQTS themselves or within their families were excluded. User group participants who posted their initial new diagnosis story more than 4 years prior to this study were excluded.

Protection of participants. The data collected in this study are available in the public domain of the world-wide-web; therefore, participants in this study did not necessarily know that their posts were included in this research study. This process is considered a best practice in internet-based research because it reduces the risk to the participants by not informing them. Informing a public community that it will be

observed for research purposes may do more harm to the members of that group than if they were never notified (Eysenback & Wyatt, 2002). Presentation of final results is presented in aggregate or is substantially altered to assure total anonymity of the user group participants. Furthermore, the mediators of the group are aware that passive-analysis of the user group for research purposes has been conducted (S. Felgoise, personal communication, March 2011).

Potential risks to participants. There are no potential risks to participants. In order to ensure anonymity of the participants, data were de-identified during the collection process and prior to analysis. Results are discussed in aggregate form, with the exception of quotes that have been substantially changed to protect participant identity. Consent was not needed because the information is available in the public domain of the world-wide-web. The project received approval from the Institutional Review Board at the Philadelphia College of Osteopathic Medicine in March 2012.

Procedure

Data Collection

The data were collected in the form of transcripts and were downloaded from the online user group and put into a word processing document. Each discussion thread was collected as an individual file and formatted to remove digital artifacts, such as “carrots” and backslashes. The file was further formatted electronically to delete redundant posts that were collected during the download process. Next, each file was searched electronically for participant user names and digitally replaced with de-identifying information. After the data were collected and de-identified, they were printed, collated and bound for analysis. A de-identified copy was provided to each member of the

analysis team. After the data were collected, they were analyzed as described in the next section.

Data Analysis

The analytic and interpretative process of grounded theory of qualitative analysis as described by Strauss and Corbin (1998), served as a foundation for data analysis. This qualitative process describes and interprets direct experience, to provide new insights and to generate theory. Grounded theory of qualitative analysis includes a consideration of variables that emerge as themes within the online user group. The process also elaborates on the complex gestalt observed as the experience of the online group (Kazdin, 2003).

Validation team. Data were analyzed through a validation team. The team consisted of three clinical psychology doctoral students, specifically trained in the grounded theory of qualitative coding methodology, under the indirect supervision of a licensed psychologist. Coders met prior to engaging in the coding process and received additional instructions and training regarding the coding strategy for this project. After initial training was complete, each team member was provided with transcripts for review and each conducted an independent coding process prior to meeting with the other members of the validation team. Each team member kept process memos to describe the analytic process and inform methods of validity and trustworthiness during prolonged engagement in the data. These notes support process of coding including the use of triangulation, searching for disconfirming evidence and reflexivity. These techniques are of value because they ensure that the process and procedures are consistent, despite changes in constructs as they emerge (Marshall & Rossman, 2011).

Coding techniques. Triangulation required the use of more than one point of data to produce greater understanding of a single construct; this technique is of value because it increases the external validity of the research findings. The process of searching for disconfirming evidence is an additional technique that was used to further elevate the interpretative analytic coding process. It is of value because it considers additional possible counter-intuitive perspectives in addition to those initially identified, and thereby included alternative interpretations of the data as part of the coding process. Additionally, the process of identifying themes included a reflexive process that repeatedly checked for information found in process notes and transcripts as concepts were formulated. Reflective analytic coding was of value because it encouraged triangulation between multiple data points and ensured consistency in procedures within changes in constructs as they arose.

Functional assessment. In addition to this process, the data were further analyzed through use of a functional assessment. That is, the antecedents, behavior and consequences were identified in each participant's posts. By identifying the antecedents, consequences and patterns of use (continuation, discontinuation), observations regarding the suspected function and purpose of the interaction are proposed. Therefore, the themes produced are functional themes that address the research question under investigation. Themes specifically examined the purpose for using the online user group when coping with a new diagnosis of LQTS. This is of value because, in addition to finding trends, the observation of the participants' behavioral interaction with the online user group provides additional information about the function and utilization of the group for each user.

CHAPTER 3

Findings

Participant Characteristics

All 9 participants had at least one child with a recent diagnosis of LQTS; all 9 were mothers and one was also a grandmother. Seven participants were mothers with no knowledge of LQTS prior to the emergence of symptoms in their children. Five of the 7 mothers with symptomatic children were also newly diagnosed with LQTS themselves. Two mothers with symptomatic children did not have any other family history of LQTS; that is, their children were considered to have a spontaneous mutation of LQTS. Two participants were mothers who knew they had LQTS prior to the diagnosis in their children. Two of the 9 participants had children who died from LQTS, one prior to their initial posting on the user group and the other after engaging with the group for several years.

Although 7 of the 9 mothers were diagnosed with LQTS, the content of their posts focused on their children's prognosis, suggesting a heightened concern with the syndrome in their offspring. This is further illustrated by the observation that all 9 participants were mothers of children with LQTS. Although the user group was searched for all 'new diagnosis' stories, it is noteworthy that the stories collected were that of participants' children, rather than of the adult user group participants themselves. This suggests that the experience of having a child diagnosed with LQTS motivated these participants to initiate their first introductory post. In fact, one member informs the group that she read posts for several months prior to her initial contribution. When she did finally post, it was the day after genetic results confirmed LQTS in 2 of her 4 children.

Table 1.

Description of Participant Characteristics

Summary of Participant Demographics

(Names and ages have been altered to protect identity)

Pseudonyms	*Age*	*LQTS Type*	*Family Hx*	*Children with LQTS*
Nadine	unknown	None	None	infant w Borderline LQTS
Anne	unknown	LQTS1	Yes	Daughter w both LQTS 1 & 3 2 other children w LQTS 1 Husband may have LQTS 3
Helen	mid 60's	LQTS 2	Yes	Daughter and granddaughter
Kristy	mid 30's	LQTS 5	Yes	2 daughters w LQTS 5
Becky	early 20's	unknown	Yes	infant w LQTS and Wolff Parkinson White Syndrome (WPW)
Peggy	early 40's	LQTS 3	Unaware	school-aged son w LQTS 3 school-aged daughter died of undiagnosed LQTS 3
Grace	unknown	LQTS 1	None	3 children suspected LQTS
Lacy	unknown	None	None	infant w LQTS 2 and Tetralogy of Fallot (ToF)
Bonnie	unknown	unknown	None	school-aged son with LQTS school-aged son EKG normal

Functional Assessment and Theme Findings

The following is based on a functional assessment that identified the antecedents and consequences of each participant's conversational contributions. By identifying the antecedents, consequences and patterns of use (continuation, discontinuation), observations regarding the suspected function or purpose of the interaction can be proposed. Although the results of the data are presented in aggregate form, isolated segments of participants' stories are offered to elucidate the richness of their experiences. Please note that identifying information has been substantially altered to maintain participant anonymity. All of the participants' and children's names, age, location and descriptors have been altered to protect participants. The functional conversational assessment revealed the following functional themes.

Seeking Connection and Normalization

One of the strongest recurring themes identified was an expressed desire to connect with others who have shared a similar experience or who may identify with participants' unique situations. All of the 9 conversations that were analyzed contained questions seeking normalization and connection. For example, questions that recurred throughout the transcripts included: "*Who has had a similar situation?" "Am I alone?" "Is my situation unique?" "Is this normal?" "Who understands?" and "Is anyone located in my geographical area?"*

Eight of the 9 participants received direct responses from peers who described at least some similar experience to that of the index participant, in effect validating their request for normalization and reinforcing their posting behavior. The one participant who did not receive a reply from a peer with a similar experience described a very unique

presentation of LQTS. Interestingly she did not continue to contribute further posts. This pattern of posting suggests that the need for normalization may be a maintaining factor for some user group participants. This participant sought normalization in her introductory post and when that need was not met by her peers, her posts discontinued.

Negative medical experience: A negative medical experience was identified as an antecedent to posting on the user group. Five of the 9 participants described various dissatisfying experiences with medical care, ranging from frustration and disappointment to difficulty with insurance, to general anxiety with their child's current treatment recommendations. There was an observable pattern in which participants posted to the user group to discuss frustration with lack of information provided at medical appointments or minimization of LQTS symptoms in the medical community. The consequences of this posting behavior included validation, confirmation and general expression of support from the online user group community. The function of this behavior, therefore, appears to be to find validation, empathy and process a difficult experience.

Positive medical experience: To the contrary, another participant narrated her positive medical experience when her pre-school aged daughter required tympanostomy tube insertions (ear tube surgery). In this example, the antecedent to the participant's behavior included expressed anxiety regarding the upcoming surgical procedure. She posted to the group, discussing her concerns as well as the actions she was considering, demonstrating a positive, rational problem-solving orientation. She later updated the group and reported a successful outcome. The consequence of her behavior was an expressed "good feeling"; she also received 2 peer replies congratulating and

encouraging her, "*Wonderful news! And think about how many people you have now educated on LQTS*!" This example illustrates a proactive approach that engendered a sense of control regarding her daughter's medical procedure.

Seeking Emotional Support

Seeking support was another prominent theme that emerged from the data analysis. Experiencing a lack of support regarding LQTS was identified as an antecedent to posting on the user group. Within their posts, several participants described an absence of emotional support from their families or communities at home. As a result of this expressed lack of support, participants received validation, confirmation and general expression of support from the online user group community. The function of this behavior then, appears to be to find validation, normalization and gain emotional support.

Helen's story. *"We are dealing with husbands who just aren't into the LQTS thing*." Helen has LQTS and is both a mother and a grandmother to a child with LQTS. After her adult daughter gave birth, the new baby was diagnosed with LQTS and her daughter's LQTS worsens postpartum. Her daughter was recommended for ICD surgery. Her granddaughter was brought home from the hospital on beta-blockers and with extra monitoring equipment. Helen took a leave of absence from her job and moved into her daughter's home to help her through the surgery and care for her infant granddaughter. Helen posted frequently during her "*tour of duty*." She reported finding support in the group during a time when she has been burdened with extra care for persons' coping with LQTS. She also described an absence of support outside of the user group.

In additional to seeking support, participants expressed a variety of emotions related to coping with a new diagnosis of LQTS. Although the majority of emotional

content focused on difficult experiences, it is noteworthy that participants also expressed thanks and gratitude related to joining the online community and for having the knowledge of an accurate diagnosis. The validation team identified the following themes of emotional expression (Appendix E contains a Rank Order List of Emotional Themes).

Fear, Worry, Uncertainty

Every participant included remarks in their posts regarding worry and concern for their children; for example, "*It's like a monster lurking inside*" and "*this makes me so uneasy*." Most expressed a fear of the unknown and used terms such as, "scared, concerned, uneasy or doubt." Fear and worry was often expressed in the early stages of diagnosis, coupled with lack of information or connection to resources. For example, "*perhaps my daughter's symptoms presented so early because she inherited something from both sides*." Anxiety and uncertainty was expressed regarding situations and triggers that are difficult to control, such as the startle response in infants. Many participants also expressed fear and anxiety while waiting for the results of genetic tests. Mothers further discussed unease around levels of monitoring and treatment, "*the boys are only monitored yearly and this makes me uneasy.*"

Interestingly, a minority of participants expressed their fear and uncertainty with an anxious urgency. Kristy for example says: *"I feel like I could lose them at any minute….I can't get them to see the EP until the 3rd … and as you know waiting is the worst part… it's like being told they have cancer, except there is no time."* Grace offered a fitting summary in her closing remarks, "*The worst part of having LQTS in the family is the uncertainty*."

Shock, Numbness, Overwhelmed

Three of the 9 participants described feelings of shock, numbness or a sense of being overwhelmed. Often these feelings were expressed in the initial stages of a new diagnosis. For example, upon hearing of her new LQTS diagnosis and learning that all 3 of her children had LQT changes on their EKG, Grace expressed a sense of being overwhelmed, " *This past month has been unreal…this is a lot of change right now I am pretty overwhelmed*." Another participant, Peggy explained that her "*head is spinning,*" as she considers the decision to go through with scheduled ICD surgery or cancel and get a second opinion. Helen expressed a great sense of shock bringing home an infant with a monitor, "*We just never thought about going home with wires, monitors and beta blockers on a tiny baby*." Later Helen expressed a sense of being overwhelmed when her daughter was unable to care for the new baby because of scheduled ICD surgery postpartum, "*things seem to have snowballed*."

Frustration, Burdened, Stress

Three of the 9 participants described feelings of frustration, stress, burden and discouragement. Helen described frustration accomplishing everyday activities; her daughter had difficulty caring for her newborn and recovering from ICD surgery: "*everything seems to be a problem for her.*" Becky described frustration with a medical experience: "*dealing with doctors is extremely frustrating sometimes because we absolutely depend on them for their knowledge and authority*." Nadine described a profound sense of frustration when her daughter's genetic report was found negative for LQTS. "I *want to be happy about these recent results and hope that this was all just a mistake…I still don't know what caused my daughter's episodes and it is so frustrating.*"

Isolation, Discounted, Invalidated

Closely related to frustration, burden and stress are feelings of being invalidated, discounted and alone. The same three participants who described feelings of burden and stress also described feelings of invalidation and isolation. Becky experienced frustration when her cardiac event was minimized and discounted by the nurse. "*I didn't exactly get the response I had hoped for. The nurse called back and said that he wasn't concerned and to call back if it happened again.*" When Becky reported her event to the nurse, she had prior knowledge of LQTS because LQTS was not new to her family. Nadine, however, had no prior knowledge of LQTS because they rushed their infant daughter to the ER after cardiac events. Nadine also expressed feelings of invalidation by the ER doctors. Although Nadine had no knowledge of LQTS, they knew something was wrong. "*Each time we rushed her to the hospital. By the time we got there, she looked fine, the doctor discounted the episodes.*"

Thankful, Gratitude

In addition to expressing other emotions, 3 of the 9 participants expressed gratitude or thanks. Peggy lost her daughter to LQTS before she was accurately diagnosed. She expressed gratitude, however, that they now have a diagnosis and they can treat their other child who also has the gene. She says, "*We are thankful that we now know what is going on.*" Kristy also expressed gratitude for the advice she received from peers, "*Thank you … I find comfort from those who have been there… I am very relieved.*" Another participant, Grace, expressed gratitude for finding the online user group. "*I am so glad to have found such a wonderful group to connect with around LQTS. Having this resource is just wonderful, I am so thankful to have found you all.*"

Concerned About New Restrictions

Two participants described frustration with the new lifestyle modifications and activity restrictions for their children. Grace expressed frustration with her son's new activity restrictions; she explained to the group that her son is very active and asks the group for information about how much activity is appropriate. Peggy also expressed similar concerns regarding activity restrictions for her son. *"He is a very active boy, and has not been told he cannot swim on the swim team in the summer, take karate anymore or join the cross country team in 6th grade.*"

Guilt

Two of the 9 participants expressed feelings of guilt. These 2 participants were mothers who knowingly carried the LQTS gene and later had children who were also diagnosed with LQTS. Mothers' expressed guilty feelings when their children inherited the LQTS gene from them. Helen, for example, expressed a profound sense of anguish related to passing the LQTS gene onto her daughter and granddaughter, *"I don't feel like a very good mother [because] I gave this LQTS to my daughter and my granddaughter…. The doctors say it was a mutation starting with me."* Peers encouraged her to let go of the guilt, "*no one would pass on LQTS knowing that it's not in our control.*" Helen replied and further explained that she has tried to let go of the guilt but then it just, "*pops back up again.…LQTS has just complicated so many things in our life*."

Seeking Information

The function of many posts included seeking and gathering of information related to LQTS. Connecting with peers to seek information was a primary function of the user group. Because LQTS is relatively rare in the general population, this unique group

served as a community for which participants could gain critical information and learn how others handled issues related to LQTS. Most of the information sought related to parenting a child or children with LQTS, but some sought limited information regarding their own needs related to a new diagnosis of LQTS. Topics of discussion included physician referrals, medication management, and surgery preparation, how to inform children of their new diagnosis and negotiating school and home visits.

Utilization

In addition to functional themes reviewed previously, the functional conversational assessment also reviewed participants' utilization trends such as, number of posts, duration of utilization, reason for initial posting, frequency of posting, duration and patterns of use.

Number of posts. A total of 198 posts were downloaded and were included in the coding process. The number of posts contributed by each participant ranged from 1 to 19 posts each, with an average of 7.7 posts per participant. The total post count included peer reply posts and participant authored threads. All participants received reply posts from peers with a range between 3 and 33 total peer replies, with an average of 14.2. (See Table 2 below)

Duration of utilization. The time that lapsed between the participants' initial, introductory post on the user group to their last post on the user group was collected and coded. The length of utilization ranged from 1 day to just under 4 years and may have continued on, past the data collection point. The rounded average duration of utilization was 369 days. It should be noted that 2 participants posted only on 1 day. That is their

first and last posts occurred on the same day, yet others continued to post over years and may have continued past the point of data collection.

Reason for initial posting. Six of the 9 participants contributed their initial post after having recently received genetic results confirming LQTS within their families. Five of the 9 participants initiated their first post while waiting for genetic results to confirm a LQTS diagnosis within their family. Three participants initially posted to the user group after experiencing a cardiac event themselves, or in their children. Three participants initially posted to the user group while preparing for ICD surgery; and two initially posted after recently undergoing ICD/pacemaker surgery. (Appendix F contains Rank Order Antecedents)

Role Reversals

As reviewed previously, the functions of the user group primarily included seeking behaviors, including seeking connection and normalization, seeking emotional support and seeking information. Interestingly, two participants demonstrated a role reversal in their interaction styles. That is, over time, it was observed that these user group participants changed from seeking behaviors to providing information and support to peers.

Table 2: Summary of Online User Group Usage

Pseudonyms	*Duration*	*Threads Authored*	*Replies to Peers*	*Total # of Posts Contributed*	*Total # of Posts Received*
Nadine:	3 months	2	1	4	9
Anne:	1 day	1	0	1	3
Helen:	2 months	5	5	19	35
Kristy:	8 months	5	2	9	12
Becky:	9 days	2	2	5	3
Peggy:	1 year	3	1	9	17
Grace:	over 3 yrs	2	0	3	10
Lacy:	under 4 yrs	5	14	19	33
Bonnie:	1 day	1	0	1	6
Mean:	***369 days***	***2.8***	***2.7***	***7.7***	***14.2***
Median:	***90 days***	***2***	***1***	***5***	***10***
Mode:	***1 day***	***3,5***	***0,1,2***	***1,9,19***	***3***
Range:	***1 day- 4 yrs***	***1-5***	***0-14***	***1-19***	***3-33***
Total Posts:				***70***	***128 = 198***

CHAPTER 4

Discussion

This study found that participants using the online user group are often parents who cope with a range of bio-psycho-social demands related to parenting children with LQTS. The bio-psycho-social demands of LQTS often occur within a medical-family-social-cultural milieu that is unfamiliar with LQTS. The additional demands of parenting a child with LQTS, in combination with the general lack of understanding of LQTS in most socio-cultural environments, contributes to the psychological distress faced by many parents who participated in the study. Findings suggest that those who choose to interact with the online user group are parents whose need for information, normalization, connection and emotional validation were not fully met elsewhere. Over time a trend was observed whereby participants' roles reversed from seeking information and normalization to providing information and emotional support to peers coping with the demands of LQTS.

Parenting children with LQTS. The main purpose of the user group was discussion of participants' children who have been newly diagnosed with LQTS. It is interesting that although the user group is designed for anyone coping with LQTS, all 9 participants selected for this study were mothers of children with LQTS. Although most of these mothers (7 of 9) also had LQTS, and 5 of them newly diagnosed, the content of their posts focused on the needs of their children rather than their own medical concerns. "*We don't see the point of me going for further evaluation before our son.*"

Previous, related works support this finding. Hendriks et al. (2005) found that parents are often more concerned about the health and welfare of their children than they

are about their own. Sharpe and Rossiter (2002) found that parenting a child with a chronic medical condition produced more negative distress for the parents than for the children. Although the previously mentioned research focused on general childhood medical conditions, Kroode et al., (2000) found that parents are often more concerned about their children's health prognosis and less concerned with their own health during genetic testing specific for LQTS. The current study supports Kroode et al., (2000) and further suggests that parents are more concerned with the health of their children throughout various stages of diagnosis and treatment.

Gonzales (2009) and Rovinsky (2011) conducted studies specific to parents coping with LQTS and also found that parents tend to focus on the medical concerns of their children rather than their own medical concerns. Based on their research, they further hypothesized that parents often unknowingly form a schema about being a "good parent," that may include core beliefs related to protecting their children and keeping them safe from harm. The nature of LQTS complicates parents' ability to maintain this schema and further challenges their beliefs about being a good parent, resulting in heightened levels of emotional distress (Gonzales, 2009; Rovinsky, 2011).

The current study provides further support for Gonzales (2009) and Rovinsky (2011) including further validation of their findings that parenting a child with LQTS is different from parenting other children, including children with other chronic medical conditions. LQTS is different from other chronic medical conditions because the triggers are everyday events such as sleep, emotional responses, startle or activity, and the consequences can be sudden and life threatening. The fact that these factors are often outside the control of parents adds to their difficulty. Psychologically, this disrupts

parents' fundamental core beliefs about their ability to protect their children from harm and greatly adds to their experiences of distress.

The current study found that parents interacting on the online user group express this distress by seeking emotional support, normalization, validation and information from peers with similar experiences. Based upon the findings in the current study, in combination with others (Gonzales, 2009; Rovinsky, 2011), it may be further suggested that LQTS creates additional psycho-social-emotional demands on parents. When these demands exceed the resources of the current bio-psycho-social-cultural milieu of the family, additional resources are needed. This study found that the main needs of participants included seeking connection and normalization, seeking emotional support and seeking information regarding parenting children of LQTS.

Seeking connection and normalization. This study found that all of the 9 conversations that were analyzed contained questions that sought normalization and connection. One of the strongest reoccurring themes that were identified was an expressed desire to connect with others who have shared a similar experience or who may identify with their unique situations. This study suggests that meeting the fundamental human needs of connection and normalization may change when newly diagnosed with LQTS. Participants in the current study, sometimes effectively, utilized the online user group to meet the needs of connection and normalization. This study found that participants often used the online user group to seek normalization and connection relative to their unique and personal experiences with LQTS.

Seeking emotional support. This study found that seeking emotional support was another prominent reason that participants interacted with the online user group.

Some participants engaged in this online user group over years and essentially created an online community to meet their increased needs for emotional support when coping with LQTS. This is particularly important during the life transition of having a child newly diagnosed with LQTS; additional demands are placed on parents within a context that is often void of emotional resources to meet those demands. It is suggested, therefore, that additional outlets for social-emotional support, normalization and validation are needed for some families coping with a new diagnosis of LQTS.

The findings from this study in combination with others (Felgoise et al., 2012), conclude that use of an online user group for LQTS may promote increased psychosocial functioning through peer support for some newly diagnosed participants. As described previously, for a minority of user group participants, however, psychosocial benefits may not result from online user group interactions. Results from this study suggest that in some cases, their distress may actually become worse. In sum, most participants received psycho-emotional benefits from use of the online group; the few who do not benefit may experience worsening symptoms of distress as a result.

Emotions during diagnosis. Anderson et al. (2008) found that people often express feelings of relief and validation when newly diagnosed with LQTS. The current study provides additional support for Anderson et al. (2008) and further suggests that when symptoms go undiagnosed for a period of time, gaining a correct LQTS diagnosis may result in feelings of relief and validation, as well as feelings of gratitude for gaining information that will protect their children. Lane et al., (2009) found that positive emotions and happiness in particular may buffer individuals from future cardiac events associated with LQTS. In light of Lane et al., (2009), experiencing the positive emotions

that may occur when a patient is accurately diagnosed may provide additional physical and emotional benefits, particularly after a period of struggle during diagnosis.

Emotions and prior LQTS knowledge. Mothers who had prior knowledge of LQTS were more likely to express feelings of guilt, as compared with mothers with no prior knowledge. This seemed to occur because mothers, who knowingly carried the LQTS gene, hoped for a chance to prevent the continuation of the gene in their children. Often this desire was simultaneously expressed with a cognitive distortion such as magical thinking or misinformation. *"Because I had been taking the medication, I thought it would be prevented in my children,"* and *"I just desperately wanted to prevent this and I feel so guilty [because] this is my fault."* This speaks to the strong psychological need for parents to protect their children from harm and when unable to do so, may lead parents to distort their beliefs and cognitions in an effort to maintain psychological homeostasis.

Emotions with older, newly diagnosed children. The current study found that participants with older, school-aged children, newly diagnosed with LQTS were more likely to express concern about activity restrictions. Two participants in the current study had children who were school-aged when diagnosed with LQTS. Both of their posts focused on concerns related to activity restrictions. The results of the current study provide further support for Farnsworth et al., (2006), which suggest that older children and adolescents may have more difficulty responding to a new diagnosis of LQTS than younger children. Older children have developed interests and hobbies such as sports, swimming, etc. They may have considered future career interests that involve a range of activities, including some interests which are commonly restricted due to LQTS.

Younger children, however, may not have yet developed these interests that conflict with activity restrictions related to a new diagnosis of LQTS.

Seeking information. This study found that participants utilized the group to seek information about LQTS. Connecting with peers to seek information was a primary function of the user group. Because LQTS is relatively rare in the general population, this unique group served as a community in which participants could learn how other people handled issues related to LQTS and gain critical information. Most of the information that was sought related to parenting a child or children with LQTS; some, however, also sought information regarding their own needs related to a new diagnosis of LQTS.

Parents described a lack of understanding regarding LQTS within the general community, within families and within the medical communities themselves, such as during trips to the ER, for example. A cultural environment that lacks information or understanding regarding LQTS contributed to feelings of isolation and lack of support from the larger family network. Support and understanding is often foundational in supporting parents who struggle when parenting an ill child. The fact that children with LQTS often do not appear ill intensifies the need for parents' coping with a new diagnosis of LQTS in their children. This fact may motivate parents to find a community that offers useful information and understanding based on their own similar experiences. The complexities of the biology behind the condition also contributed to parents' lack of understanding regarding the nature of LQTS. Parents utilized the online user group to gain this information.

Parents also sought information regarding strategies to cope with every day stressors related to LQTS. Parents often lack the ability or information to moderate the factors that contribute to sudden cardiac events in the children because triggers are every day stimuli that parents often have little or no control over. For example, one participant asks, "*Does anyone know how to keep a baby from startling*." Participants sought information about a myriad of concerns regarding strategies for managing ICD surgery, to preparing for doctors visits and negotiating school and community activities. Participants also discussed how and when to inform their children of their LQTS diagnosis.

The current study supports findings from Anderson et al., (2008) and Hendricks et al., (2005), which suggest that those newly diagnosed would benefit from regular and sequential information regarding LQTS. The current study further suggests that a peer based, online format is a method for receiving and providing this information. Also, there are additional benefits that are often garnered from peer-provided information. Over time, users may shift roles from seeking information to providing information, keeping them connected to a community coping with LQTS. This prolonged engagement provides resources to members of the community such as role models for coping with LQTS at various stages of diagnosis, treatment and medical management.

Relationship to Previous Works

To date, the current investigation is the only study that has specifically examined an internet-based user group to measure the function and utilization patterns of those coping with LQTS. Felgoise et al. (2012) presented an abstract at The Heart Rhythm Society (2012) regarding the psychosocial difficulties related to living with LQTS that

included the aforementioned research on the LQTS user group. These findings, absent from the current study, conclude that collective challenges include a lack of general knowledge regarding the medical condition, having medical expectations that exceeded perceived experiences during initial diagnosis, emergency room visits during ongoing medical treatment. Felgoise et al. (2012) also found that the user group offered support for participants struggling with these challenges of LQTS. The current study offers additional validation of the findings of Felgoise et al. (2012) and provides further elucidation regarding the function and utilization of the online user group for participants coping with the psychosocial challenges of LQTS.

Terror management health model (TMHM), introduced by Goldenberg and Arndt (2008), is a theory that describes the role of death in one's health behaviors. TMHM proposes that mortality awareness and self-esteem are critical determinants of health related thinking and behavior. TMHM describes what motivates people relative to their health choices and behaviors; TMHM describes both positive and negative health behaviors relative to individuals' level of mortality awareness and their desire to conform to society expectations (Cox et al., 2011). Regarding smoking behavior for example, TMHM suggests that the degrees to which an individual's self-esteem is contingent on smoking and the degree to which mortality is salient, will influence smoking behaviors. Specifically, if the person believes he or she can quit and that doing so is important for longevity, he or she is more likely to quit smoking (Hansen, Winzeler & Topolinski, 2010). Regarding the current study, TMHM may provide further analysis of the descriptive findings in the current research, relative to parents' choices related to managing their children's LQTS.

By nature of the current study, participants were engaged in active coping by posting to the online user group to gain information, normalization and emotional support. In addition, participants in the current study were engaged in health promotion and maintenance behaviors primarily for their children and sometimes for themselves. Interpreting active posting behavior from the TMHM, researchers found that participants in the current study arguably have: (1) developed self-esteem related to active posting for the benefit of themselves and/or their children and (2) activated mortality salience, given the nature of LQTS, which may have been a sufficient condition in motivating their behavior to post.

TMHM may also describe the behavior of passive user group members. Passive users are those members who join the group and may read the posts, but do not actively contribute written posts to the online user group. Researchers, interpreting behavior from the TMHM, believe that passive posting behavior of the participants in the current study suggests that (1) they have not developed the self-esteem to believe that their written contribution to the group will benefit themselves or their children and/or (2) their belief about the nature of LQTS has not activated their mortality salience and/or (3) the behavior of joining the group and passively reading the written contributions of others is sufficient, which precludes the necessity to post.

Clinical Implications

This study suggests that parents who use an active coping style believe that their child is at risk due to LQTS and believe that the act of contributing posts to an online user group may benefit their child; these are the parents who are most likely to benefit from joining an online user group as investigated in the current study. Parents who are seeking

information, normalization and social connection around the issues related to parenting a child with LQTS, may also benefit from the user group. Parents who do not believe that their child is at risk for sudden death or who do not believe that the action of joining a group might help their child are less likely to benefit from use of the online group.

Limitations

This study searched the user group for new diagnosis stories that have been recorded within the previous 4 years and produced only 9 participants. Given the fact that LQTS occurs 1 in 2,000 births (Schwartz et al., 2009), it is clear that many newly diagnosed patients never post to the online user group. By the nature of this study, information about the psycho-emotional needs of those individuals is still unknown. Findings from this study do not generalize to all people newly diagnosed with LQTS; rather, it contains information gleaned from those who choose to actively post to an online user group.

Generalization of the data is also limited because of the archival nature of the data set. Demographic information regarding age, gender, sex, race, ethnicity, nationality, regional location and religious preferences were limited to that which has been arbitrarily provided on the user group. The archival nature of the study further limits the information collected to answer the research question under investigation. The investigator was unable to interview the participants; therefore, specific questions about the purpose and use of the user group were limited to those patterns observed through functional assessment and qualitative theme analysis. Although the data collected spanned a four year time period and provided additional information about those interacting in the user

group over time, information about those who discontinued its use was limited to the patterns observed in the functional assessment.

In addition, the nature of this study included those participants with access to a computer and the internet, further limiting inclusion of groups with barriers to this resource. Groups prevented from inclusion, based on lack of computer and internet access may include low socio-economic groups as well as very young or very much older persons who lack computer skills. Based upon these limitations inherent in the research design, generalizations of the results are limited and conclusions are theoretical rather than causal in nature.

Suggestions for future work

Further research is needed regarding the psychosocial challenges of coping with LQTS. Research in this field is in its infancy; it is minimal and is largely exploratory in nature, compared with research on other medical conditions of childhood. Early research suggests a range of psychosocial difficulties, including lack of understanding, frustration with medical care as well as anxiety regarding treatments such as surgery and activity restrictions (Felgoise et al., 2012). Furthermore, recent research suggests that positive emotions, happiness in particular, may buffer individuals from future cardiac events associated with LQTS (Lane et al., 2009). More research is warranted.

Future research may include further investigation into the findings proposed in the discussion of the current study. Discussion points generated from the current study that warrant further analysis include: (1) meeting the fundamental human needs of connection and normalization may change, based on the lack of LQTS models in society, (2) parenting a child who is at risk for sudden death may challenge parents' core beliefs

about being good parents, increasing psychological conflict, (3) utilization of an online user group for LQTS may help to support healthy emotional coping and support peer based coping and (4) LQTS may create additional psycho-social-emotional demands on parents, compared with other childhood medical conditions. When these demands exceed the resources of the current social-cultural milieu of the family, additional resources are needed. If further qualitative research supports these theories, a quantitative analysis is suggested.

Another suggestion for future research is to interview newly diagnosed patients who choose to participate in a user group and also those who choose not to participate in an online user group to determine, further, the psychosocial benefits and drawbacks of such online user group interaction. Further research is also needed to compare other methods for meeting the psychosocial needs of families with LQTS. In addition to the unstructured informal user group used in the current research, other forums warrant investigation; these include peer led, in person groups, professionally led therapeutic support groups, structured formal online user group, peer dyads for information-emotional exchange and so on.

Based on the findings in this study, public role models and examples are needed for the LQTS community. A model of positive parenting for children of LQTS would be a welcomed resource for those struggling with the spectrum of parenting stages from new diagnosis to relinquishing medical care as children leave the nest. Additional research and publication efforts are needed to meet the needs of the LQTS community, including needs for practical information, normalization, validation and emotional support as well as advocacy and social outreach efforts to educate the general public regarding LQTS.

Summary

LQTS is a sudden death syndrome in which persons may live asymptomatic for years and then die suddenly upon initial presentation of symptoms. LQTS also affects a large percentage of the population, with recent estimates suggesting its occurrence in 1 in 2,000 births (Schwartz et al., 2009). LQTS is caused by genetic mutations that affect the electrophysiology of the heart, resulting in a prolonged QT interval, electrical instability, resultant ventricular arrhythmias and sudden cardiac arrest and death. Treatments include medications, activity restrictions, implantable cardioverter defibrillators and left cervical sympathetic denervation in selected cases. Little research exists regarding the psychosocial factors of the illness or how families cope with the management of the chronic illness. The current study used archival data from an online user group. Participants were members of the group who discussed concerns related to LQTS. The aim of the current study was to investigate the function and utilization of the online user group while coping with the psychosocial challenges of living with LQTS. The study contributed to a number of prior researches which investigated the online user group for themes related to anxiety (Janney, 2011), compliance (Steinhauser, 2010) parental control (Rovinsky, 2010) and grief and loss (Haynes-Weller, 2011). The current study was the first to specifically examine the function and utilization trends of the online user group.

The current study found that all 9 participants selected were parents of children newly diagnosed with LQTS. This study found that the main needs of participants included seeking connection and normalization, seeking emotional support and seeking information regarding parenting children of LQTS. Over time, utilization patterns shifted from seeking behaviors to providing behaviors to peers. In essence, participants engaged

in this online user group to create a community to meet not only their needs for emotional support, but also practical information when coping with a new diagnosis of LQTS.

The findings of the current study, in combination with findings from previous studies (Felgoise et al., 2012), support results which suggest that utilization of an online user group for LQTS may encourage healthy emotional coping. Benefits of the online user group include an approach-based emotional coping behavior, in combination to connection with peers who have similar experiences. Regarding new diagnosis, Anderson et al. (2008) found that people often express feelings of relief and validation when newly diagnosed with LQTS. The current study provides additional support for Anderson et al. (2008) and further suggests that when symptoms go undiagnosed for a period of time, gaining a correct LQTS diagnosis may provide feelings of relief and validation, as well as feelings of gratitude for gaining information that will protect their children. In addition, mothers who had LQTS were more likely to express feelings of guilt when a child was diagnosed, as compared with mothers with no prior knowledge of the disease. This seemed to occur because mothers who knowingly carried the LQTS gene had hoped for a chance to prevent the continuation of the gene in their children. The current study also found that participants with older, school-aged children, newly diagnosed with LQTS were more likely to express concern about activity restrictions. Regarding user group utilization trends, this study found that over time individuals' psychosocial needs might change from seeking information and emotional validation to those of providing information and support for others in the LQTS online user group community. Further research comparing additional resources for meeting such needs are warranted. With further research, results from this study may generalize to other types of

groups, such as in-person, professionally led adjustment or therapy groups, peer led support groups or other group information sessions. Advocacy efforts to educate the general public about LQTS may lessen the burdens of a new diagnosis and of managing ongoing care.

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Appendix A

Comprehensive Literature Review

Theoretical Underpinnings

Coping Process

Definition of coping process. Coping is any attempt to control, minimize or prevent stress. Based on cognitive- behavioral theory, coping is an individual process that includes an appraisal of the demands of an environmental situation and personal factors based on an individual's life experiences such as their values and beliefs (Lazarus, 2000). Coping style is therefore a combination of an individual's primary perception of the situation in combination with their past experiences and acquired skills. This process is flexible and evolves over time, as the individual's unfolding life experiences alter his or her appraisal. The initial appraisal of the situation is a main determinant in how well an individual ultimately copes. According to Lazarus and Folkman (1984), there are four initial appraisals that an individual may make in any given situation. The situation may be perceived as (1) benign, in which case no additional coping is needed, (2) harmful or involving a loss, (3) threatening or (4) as a challenge, in which case various coping styles may be employed. Although various conceptualizations exist for coping strategies, Aldwin and Yancura (2004) suggest that there is agreement within the field regarding the five types of coping strategies; (1) problem-focused coping, (2) emotion-focused coping, (3) social support, (4) religious coping and (5) meaning-making or cognitive re-appraisal.

Problem-focused coping. Also called instrumental action, this type of coping includes action-oriented ways of thinking and behavioral acts aimed at solving the problem. Problem-focused strategies may include seeking more information, taking

direct action, or breaking the problem down into manageable steps or phases, known as 'chunking'. Adaptive problem-focused coping may also include suppressing or withholding action as an effective way to cope with difficulties. For example, waiting until more accurate information is available prior to acting may result in a better outcome, as may delaying a conversation rather than acting in anger (Aldwin & Yancura, 2004).

Emotion-focused coping. This style of coping involves ways of thinking and behaviors directed at managing the emotional response to the problem. Various emotion-focused strategies exist, such as avoidance, withdrawal, expressing emotion and disengagement. Also, the use of substances such as alcohol, drugs or food may be a strategy used to cope with the emotional responses to various situations. Some emotion-focused coping is associated with poorer outcomes, such as avoidance, withdrawal and use of substances (Aldwin & Revenson, 1987); however, other emotion-focused strategies such as journaling are related to better outcomes (Aldwin & Yancura, 2004).

Social support. This coping behavior involves elements both of problem-focused and of emotion-focused coping strategies. Seeking support from others may include asking for advice, requesting emotional encouragement or justification for one's actions or ideas (Thoits, 1986). The outcome of seeking social support is generally positive, unless the respondent reacts negatively. Seeking social support can increase a person's emotional distress as a result of seeking social support and then receiving critical or negative feedback as a result (Aldwin & Yancura, 2004). Therefore, it seems that the feedback from others depends on a person's perception and appraisal of the interaction. Despite these potential drawbacks of seeking social support, it has been found to be a

protective factor for most people, particularly those from a well-functioning family, when facing challenging stressors, such as chronic illness (Dobbie & Mellor, 2008).

Religious coping. This coping behavior may also include aspects both of problem and of emotion-focused coping as well as elements of social support. Religious coping involves activities such as, prayer, concrete aid, emotional support, and asking for advice. Religious coping may include attempts to reframe difficult experiences into a positive meaning-making experience. Although the study of religious coping is relatively new, studies have found that it may be most helpful when stressors are uncontrollable (Aldwin, 1994) or when coping with chronic situations, such as caring for others, particularly in lower socioeconomic groups (Cupertino, Aldwin & Schulz, 2000).

Meaning-making. This last category, meaning-making, is sometimes called reframing because it involves a process of actively looking for the positive or meaningful aspects of an otherwise difficult situation. Meaning-making includes strategies such as "looking for the silver lining," or trying to find a benefit or positive outcome to the situation. Meaning-making may be particularly helpful when coping with intense, chronic stressors, such as illness, death or loss (Mikulincer & Florian, 1996). For example, when faced with a terminal diagnosis of cancer, an individual may, over time, come to view his or her life as ever precious and redirect his or her energy toward making amends and fulfilling benevolent life wishes and perhaps even give recognition to the adversity in life for giving a direction toward a higher path.

The manner of coping in which one engages has been linked to various health outcomes. The fact that the process of coping is dependent on how the individual appraises a situation means that the experience of the event is largely dependent upon

how that individual thinks, rather than the particulars of the situation itself. This means that people may have more control over their experiences than they perhaps exercise. Being able to cope effectively with the variety of life's inevitable difficulties is an exercise in psychological control; therefore, doing it well may be a marker of psychological health. On the other hand, maladaptive coping styles may lead to breakdowns in psychological functioning and contribute to symptoms of stress, anxiety and depression.

Social problem-solving and coping with LQTS

Social problem-solving defined. Social problem-solving is the cognitive-behavioral process of solving problems in the environment, as they occur naturally (D'Zurilla, Nezu & Maydeu-Olivares, 2004). The term social is used to denote the fact that problem-solving can be applied to any type of social interaction in which there is a problem and emphasizes the connection between problem-solving and functional adaptive outcomes. This may include, impersonal problems (finances, damaged property), intrapersonal problems, (thoughts, feelings, behaviors and health) and intrapersonal problems (martial conflict, work-place problems). The social problem-solving model may also be applied to broader socio-economic, political or community based problems, such as crime or social injustice (D'Zurilla, Nezu & Maydeu-Olivares, 2004). Therefore, social problem-solving can be used to study all types of problems, including medical conditions such as LQTS, that may affect an individual or groups of individuals,.

Similar to the general coping process as described in the previous review, the implementation of social problem-solving varies in style and skill level. Not all styles or

implementation efforts result in universally beneficial outcomes. Social problem-solving is a multidimensional process that requires both *solution generation* and effective *implementation* of those solutions. These two skill sets are not always correlated; solution generation may be applied across settings, whereas solution implementation will need to be modified to fit the specific environmental situation. Therefore, individuals may possess adequate solution generation skills, and lack necessary implementation skills or vice versa. Individuals who have, however, learned to apply effective problem-solving skills are more likely to reap the benefits of successfully navigating social and environmental barriers. Effective social problem-solving is more likely to lead to decreased stress, anxiety and associated discord. Problems left unresolved or ineffectively solved, however, are more likely to result in increased frustration, stress, negative mood and maladaptive physical and psychological distress (D'Zurilla & Nezu, 2007). Therefore it will be important to explore, identify and understand the problem-solving skills and styles of individuals who interact with an internet based user group as they cope with LQTS. D'Zurilla, Nezu and Maydeu-Olivares's (2004) model of social problem-solving will be offered as a framework to explore how individuals are interacting with and utilizing an online user group as they cope with LQTS during the first year post diagnosis.

Dimensions of social problem-solving

Components of social problem solving. Social problem-solving is a multidimensional process which initially considers two separate but interrelated components (1) problem orientation and (2) problem-solving skills. *Problem orientation* is the set of relatively stable, automatic thoughts that a person has when encountering a

problem. These automatic thoughts reflect a person's underlying cognitive-emotional schema related to the problem and may represent deeper fundamental beliefs, rules, emotions and sense of self-efficacy related to the problem. Often a person may not be fully aware of these automatic thoughts or the underlying schema motivating them (D'Zurilla, Nezu & Maydeu-Olivares, 2004). The second component is *problem-solving skills*; these are the cognitive and behavioral actions that a person may engage in an attempt to understand and solve the problem. The model describes four problem-solving skills (1) problem definition, (2) production of multiple solutions, (3) decision making and, (4) solution implementation and evaluation (D'Zurilla, Nezu & Maydeu-Olivares, 2004).

Types of problem-solving orientation. There are two types of problem-solving orientations: (1) positive or constructive and (2) negative or dysfunction. *Positive problem-solving orientation* is a cognitive-frame that includes viewing the problem as a challenge to be solved and believing that he or she possesses the ability to solve the problem with time, effort and commitment. *Negative problem-solving*, on the other hand, is a cognitive view that the problem is a threat to his or her well-being, and a doubt in his or her ability to solve the problem effectively. A person with a negative problem-solving orientation becomes easily frustrated, discouraged and avoids persistent action in the face of obstacles, whereas, a person with a positive problem-solving orientation tends to persist and maintain or evolve the action to cope with the problem effectively (D'Zurilla, Nezu & Maydeu-Olivares, 2004).

Problem-solving styles. Whereas problem-solving orientation is a more stable personality trait, problem-solving style is a set of skills that the individual employs as he

or she attempts to solve the problem. The three types of problem-solving styles described are: rational, impulsivity-carelessness and avoidance. *Rational problem-solving* is a productive style that includes a logical, purposeful, and structured application of the four problem-solving skills described previously (D'Zurilla, Nezu & Maydeu-Olivares, 2004). *Impulsivity-careless style* is a set of ineffective attempts to implement problem-solving skills characterized by limited alterative solution generation, impulsive implementation of poorly considered ideas and unsystematic review and monitoring of the process (D'Zurilla, Nezu & Maydeu-Olivares, 2004). *Avoidance* problem-solving is another maladaptive coping style characterized by postponement, inactivity and, reliance on others. Persons implementing an avoidant problem-solving style tend to avoid acting on problems and prefer to wait as long as possible, in an attempt to allow problems to resolve without action on their part or perhaps in an effort to encourage others to act on the problem for them (D'Zurilla, Nezu & Maydeu-Olivares, 2004).

Social Problem-Solving Process.

In sum, the model described by D'Zurilla, Nezu and Maydeu-Olivares, (2004) includes two possible problem orientations (positive, negative) and three problem-solving styles (rational, impulsivity-carelessness, avoidant). The model predicts that the most effective outcomes result from a combination of a positive problem-solving orientation and a rational problem-solving style. These problem solvers are more likely to have a combination of positive cognitions and behaviors that encourage active, persistent efforts, resulting in beneficial outcomes. Juxtaposed are poor problem-solvers whose combination of negative cognitive orientation and ineffective behavioral skill set result in

lack of persistence, lack of effort, avoidance or dependence on others to act (D'Zurilla, Nezu & Maydeu-Olivares, 2004).

Review of Current Research

Coping with Daily Stressors

In addition to considering the impact of an individual's previous life experiences, personal values and problem-solving style, the type of stressor may also influence the coping process. According to Aldwin and Yancura (2004), there are a number of identifiable characteristics associated with coping with everyday life stressors. They suggest that there is a relationship between the ability to control the stressor and an individual's ability to cope effectively with the stressor. Problem-focused coping is more effective when stressors are controllable, such as problems at school; whereas emotion-focused coping is more effective when dealing with uncontrollable problems, such as chronic illness. Researchers have found that having a positive problem orientation as described in social problem-solving predicted active coping and positive outcomes when coping with daily stressors (Chang and D'Zurilla, 1996). In fact, Nezu, Nezu & Lombardo (2004) suggest engaging both in problem and in emotion focused coping for healthy outcomes when challenged with stress.

Additionally, the benefits of coping with daily stressors vary across time (Aldwin & Yancura, 2004). For example, avoidance coping techniques may initially improve psychological distress; however, over time it may engender greater distress if a significant problem remains festering without resolve. Avoidance strategies have pitfalls such as these; however, they can offer a much-needed reprieve from stressful situations, thereby allowing an individual to rest and refocus energy towards more adaptive coping

strategies. Coping is an individual process; strategies that work for one individual may not work for others. Differences in effective coping strategies may be due to a myriad of factors, such as one's preexisting mood state. For example, individuals who are depressed seem to cope in ways different from those who are not depressed. In addition, the skill level of an individual may need to be considered. For example, problem-focused, direct action strategies may require additional skills that have not been acquired or the individual may need additional resources to implement particular strategies. In sum, the process of coping with every day stressors involves a complex interplay between the situation and the individual's values, beliefs, abilities, skills and resources.

Coping with Trauma

Research suggests that the process of dealing with traumatic events differs from the process of coping with everyday life stressors. A traumatic stressor can be defined as an event in which a person's life is in danger or a loss is threatened or endured. Aldwin (1999) found that individuals report experiencing no control over their thoughts and behaviors during the traumatic event. This is an important finding because, as described previously, the process of coping includes one's thoughts and behaviors regarding the event. Other research has indicated that disclosure of the trauma is associated with better short-term and long-term outcomes (Smyth, 1998); however, the response of others to the disclosure may mitigate these benefits. If the respondent is negative, the outcome of disclosure may be worse than if the trauma had never been disclosed at all (Silver, Holman & Gil-Rivas, 2000).

Meaning-making after traumatic experiences may include a process of accommodation and assimilation as the individual struggles to incorporate the trauma

into his or her worldview. Some researchers have found that meaning-making may be associated with personal growth after trauma. Schnurr, Rosenburg and Friedman (1993) found that men in combat experienced a change in their Minnesota Multiphasic Personality Inventory (MMPI) scores from pre to post combat. Men who experienced moderate levels of combat were found to have the most improved MMPI scores, as compared with those who experienced heavy combat or no combat.

Coping with Chronic Medical Conditions

Coping with a physical injury or a medical event can be considered a traumatic experience because it threatens to cause physical harm or even death; whereas, coping with chronic medical conditions can be viewed as daily hassles or stressors. Research regarding people's ability to cope specifically with chronic medical conditions expands the concepts of coping to consider psychosocial functioning in general. Regarding childhood medical conditions, the psychological impact of chronic daily stressors often extends from the patient to include the whole family unit. Therefore, when considering chronic medical conditions of childhood and adolescence, the psychosocial well-being of the whole family is to be considered.

Thompson and Raezer (1998) conducted a systematic review of the existing research regarding families of children with chronic medical conditions and found that epidemiological surveys conducted in the late 1960s reported that approximately 30% of children with chronic medical conditions were likely to experience maladaptive psychological functioning (Pless & Roghmann, 1971; Cadman et al., 1987 as found in Thompson & Raezer, 1998). Thompson and Gustafson (1996), however, found no relationship between chronic medical conditions and psychological impairment among

the siblings of children with chronic illnesses. Thompson and Raezer (1998) conclude that the impact of a chronic illness diagnosed in childhood is significant; however, the majority of children and families are resilient despite these demands and are able to adaptively function with a chronic medical condition.

More recently, Barlow and Ellard (2006) conducted a systematic review of the current literature, including all recent meta-analyses regarding the psychosocial well-being of children and families coping with chronic illness. Results from the meta-analyses indicated that although most children with chronic illness did not present with any clinical symptoms, they were at a mildly elevated risk for psychological distress. Houtzager et al. (1999) conducted meta-analyses and found that siblings of children with chronic illness displayed an increased risk of behavioral problems. Sharpe and Rossiter (2002) found that parents reported more negative effects than their children regarding their symptoms of distress. These findings suggest that siblings and parents suffered, as much or more psychological stress than the child who was affected with the chronic medical condition. Barlow and Ellard (2006) conclude that upon review of the available research, the evidence base needs to be extended regarding the psychosocial needs of families coping with various childhood chronic illnesses.

Coping with LQTS

Although research has been conducted on the psychological adjustment and coping process related to the demands of living with general medical conditions of childhood and beyond, little to no research had been conducted specifically on the psychosocial factors of LQTS. Unlike other chronic medical conditions of childhood,

symptoms of LQTS range from asymptomatic to death which can occur rapidly and with little warning.

Coping with genetic testing. Hendriks et al., (2005) studied the psychosocial health of parents of children who were undergoing diagnostic genetic testing for LQTS. Their findings indicated that 50% of parents who were told that their child tested positive for LQTS mutation presented with high clinical symptoms of distress. After a period of 18 months, only 30% of the parents continued to indicate high levels of distress. Hendriks et al., (2005) suggested that patients' access to sufficient information as well as connection to resources may significantly reduce their distress. The authors recommend that parents of children with LQTS be provided with sufficient information, such as periodic research updates and new developments in the area of LQTS. The authors also suggest that health care providers, including general practitioners and regional hospital cardiologists, regularly become familiar with periodical reports of clinical research findings and pass this information on to patients and families coping with LQTS (Hendriks et al., 2005).

Farnsworth et al., (2006) conducted a qualitative study examining the experiences of 31 parents who were having their children tested for LQTS. Parents were more likely to report fears regarding their child's death than of their own deaths. Parents also reported frustration with the limited knowledge of LQTS among health care practitioners. Ten Kroode et al., (2000) also used qualitative methods, both clinically and diagnostically, to assess family members who tested negative for LQTS. They found that tested individuals did not respond with relief, but instead continued to express anxiety and worry outside of the hospital setting with regard to their affected relatives.

Coping with the initial diagnosis of LQTS. Regarding the age of diagnoses, Farnsworth et al., (2006) found that adolescents who were newly diagnosed with LQTS had greater difficulty adjusting than those who were diagnosed with the chronic medical condition earlier in childhood. Older children may have more difficulty adjusting because of sudden limitations placed on activities that they have been engaged in or had pervious enjoyed, such as sports. These limitations may be recommended along with the new diagnosis; children diagnosed earlier in life may have lived with these limitations their entire short lives. Older children may also have possible future goals or aspirations that may have had time to develop through childhood, such as being a professional athlete for example.

Andersen, Oyen, Bjorvatn and Gjengedal (2008) interviewed seven individuals who had recently been diagnosed with LQTS to gain a greater understanding of their daily hassles, coping strategies and health care experiences. Participants expressed initial feelings of relief upon learning of their diagnosis and feelings of validation when receiving an explanation for the severe symptoms they had experienced. Patients also expressed frustration with avaible but inadequate health care information. The authors also reported that early and gradual information was the preference of the patients. The authors conclude that LQTS does have a significant impact on patients' daily lives and their families; however, patients generally learn to cope well and display overall healthy psychological functioning. Furthermore, patients' main concern was not with his or her own health or possible death but rather with that of their future generations (Andersen et al., 2008).

Coping with fear and anxieties of LQTS. Giuffre, Gupta, Crawford and Leung (2008) gave two child objective measures and two parent objective measures to 40 children and their mothers to investigate and compare children who had asthma with children who had LQTS on measures of anxiety and medical fears. The authors reported that children with LQTS had significantly more internalizing problems, such as fear of failure and criticism and were less likely to report feelings or share their feelings with others. This study also reported that mothers of children with LQTS had significantly higher levels of anxiety when compared with mothers of children with asthma. The authors conclude that the fear and uncertainty in LQTS can be tremendous for children and their mothers and that this may lead children of LQTS to suppress their fears and anxieties (Giuffre et al., 2008).

Arafa, Zahner, El-Dowaty and Moneeb (2008) conducted a cross-sectional study of 400 parents of children with heart disease in two Egyptian hospitals to examine the health related quality of life of parents who have children affected by heart disease. The authors reported several factors associated with the parent's health related quality of life; these included the child's illness severity, the type of heart disease, and the age of the child. Furthermore, the number of children in the home, financial concerns and any comorbid diagnoses of the affected child were found to be significant factors for quality of life. Female children, younger children and those diagnosed with rheumatic heart disease had parents with healthier quality of life measures. The authors suggest that practitioners should assess parent's psychological status and available social support when considering treatment recommendations for their children (Arafa, et al., 2008).

Benefits of positive emotions. Lane, Reis, Peterson, Zareba and Moss (2009) interviewed 38 patients with type 1, 2 and 3 LQTS one year-and-a-half after a cardiac event or ventricular arrhythmia had occurred. The authors reported that happiness and stress may alter a patient's susceptibility to future LQTS cardiac events, reporting that happiness is correlated with a decrease in the risk of a future cardiac event in patients with LQTS. Stress was found to have the reverse effect, increasing the likelihood of patients having another cardiac event. This study is the first to report that positive emotions may have a preventive effect on potentially fatal ventricular arrhythmias. These findings give further credence to the benefits of investigating the role of psychosocial factors including stress, coping and emotion, in treating patients with LQTS.

Seeking Support Online, Using the Internet to Cope

As our nation continues to experience increasing issues in accessing adequate health care, access to the internet for the general public continues to rise. Health-related uses for the internet have included searching for information regarding illness, fitness and nutrition, medications, researching physicians and hospitals, as well as seeking online support by joining informal medical discussion groups. Forkner-Dunn (2003) has discussed the pivotal role of the internet in the delivery of health care. He pointed out that, "The United States health care system is an outdated model in need of fundamental change. As part of this change, the system must explore and take advantage of the potential benefits of the "e-revolution," a phenomenon that includes everyday use of the internet by the general public" (p.1). With over 100 million Americans accessing the internet to obtain health related information, the internet as a tool for health care delivery,

patient self-management, patient satisfaction and health outcomes is an area that is currently underutilized and requires further research and development (Forkner-Dunn, 2003). With reports such as these, research in the area of health related internet use is burgeoning. Various Internet-based communications for managing chronic illness exist and vary in formality, structure and moderation.

Recent studies have found that the internet was being used to access information and gain support for persons coping with LQTS (Haynes-Weller, 2011; Janney, 2011; Rovinsky, 2010; Steinhauser, 2010). These studies examined an internet based user group for themes of grief and loss (Haynes-Weller, 2011), anxiety (Janney, 2011), parental concerns (Rovinsky, 2010) and compliance (Steinhauser, 2010). Research is sparse that specifically examines the role of the internet for families coping with LQTS. Several researchers, however, have explored the use of computer technology for adults and adolescents with other chronic medical conditions (Esyenback, Powell, Englesakis, Rizo & Stern, 2004; Johnson, Ravert & Everton, 2001; Kalichman et al, 2003; McCormack, 2010; Rains & Young, 2009; Salzer et al, 2010; Whittemore, Grey, Lindemann, Ambrosino & Jaser , 2010) .

Benefits of Internet Use to Cope with Chronic Illness

Although numerous computer mediated support groups (CMSG) have been created for a myriad of chronic health problems for adults, researchers are just beginning to explore this technology to reach adolescents coping with chronic illness. Whittemore et al. (2010) developed an internet coping skills training program for adolescents with Type 1 diabetes at Yale University. The authors reported that the development and implementation of a formal online training program was complicated and required

several versions during research and development of the online training tool. Twelve adolescents were included in the pilot study. Results suggest that the use of the internet as a medium to provide social support to manage chronic illness such as Type 1 diabetes is an achievable and practical format (Whittemore, et al., 2010).

Johnson, Ravert and Everton (2001) developed a CMSG for adolescents with cystic fibrosis (CF) at the Johns Hopkins CF clinic. They followed 18 teenagers over five months as they used the web-based support group from home on a computer or via Web TV access. Prior to participation in the study, 80% of participants reported that they did not have friends with CF with whom they could relate. After participating in the CMSG, members who used the site regularly reported that they had more friends who could understand their experience. Based on the authors findings, they recommended, daily attention to CMSG sites to change banners, start new conversations and provide technical management support. Moreover, parents of the teens who participated in this study suggested a parent internet-based support group for CF (Johnson, Ravert & Everton, 2001).

Kalichman et al. (2003) conducted the first study of its kind investigating the association between internet use, coping and social support for people with HIV/AIDS. One-hundred and forty-five participants completed a series of measures related to computer and internet knowledge, experiences, demographics, health, coping styles and social support. Results suggest significant correlations between disease related knowledge, active coping, information seeking and social support with health-related internet use (Kalichman et al., 2003).

Moderating Factors of Computer Mediated Support Groups

Rains and Young (2009) conducted meta-analyses of 28 studies investigating the health-related outcomes of CMSG and found that benefits varied among several variables. CMSG that consist of education and peer communication were more often associated with an increase in perceived social support, quality of life, and self-efficacy as well as a decrease in scores on depression scales. The results further suggest that several variables moderated positive outcomes. First, the group size was a moderator; that is, the larger groups provided increased access to knowledge and resources. The accuracy of information is increased with the size of the group because members are more likely to correct misinformation when the group is large, rather than allow incorrect information to go unchecked or uncorrected (Rains & Young, 2009).

Second, the available mechanisms for communication such as chat rooms, discussion boards and access to private e-mail were also found to moderate health benefits. These various types of communication technologies offer different features such as increased anonymity and reduced social cues. For example, two commonly available types of synchronicity offered in online communication channels allows users either to think carefully about and edit their responses prior to posting (asynchronous) or post immediately with the benefit of immediate feedback (synchronous). The authors suggest that those CMSG that provide access to both asynchronous and synchronous methods of online communication are likely to provide the greatest benefits for group members (Rains & Young, 2009).

A third factor related to the outcome benefits of CMSG is the frequency with which participants interact with the group. The consistency of interaction with the group,

either through a written posted contribution or through a non-observable interaction such as reading posts without posting a reply, is related the outcome for the user. Some perceived benefits of CMSG include the increased flexibility of interacting anytime and anywhere; this may actually result in less frequent and less reliable interaction with the site among some members and therefore reduce the benefits of site use. If persons do not interact with the group, they cannot access the benefits of the CMSG (Rains & Young, 2009). These findings suggest that the flexibility of online platforms is attractive to user members; however, regular commitment to site interaction is necessary for benefits.

Last, a moderating variable of CMSG includes the duration of time during which members participate in the group. Applying Walther's (1992) social information processing theory to internet interactions would imply that people interacting through computer technologies can attain the same levels of interpersonal relationship development as compared to those interacting in face-to-face interactions. Relationships built in an online environment, however, take more time to mature as compared with in-person interactions. According to Walther's (1992) social information processing theory, it would take longer for group members to develop the trust necessary to interact at a beneficial level in a CMSG. The authors conclude that the benefits of internet-based social support groups are likely moderated by the available structural characteristics of such groups, such as the number of members, the frequency, duration and types of member interactions (Rains & Young, 2009).

Drawbacks to Using Computer Mediated Support Groups

Although there are benefits and moderating factors to CMSG, research has also highlighted some problems inherent in an online support group. Salzer et al. (2010) conducted a randomized controlled trial, examining the outcomes of internet-based peer interactions for people coping with breast cancer. They found that participants tended to do worse over time on measures of psychological distress and quality of life. The authors reported that some of the peer-to-peer interactions appeared to perpetuate fear and anxiety after several participants shared with the group that their cancer had spread. Salzer et al. (2010) conclude that further research is needed to understand the effectiveness of these various types of internet-based user groups. It is important to note that the disease process of cancer is different from the chronic medical condition of LQTS. Therefore the results found by Salzer et al. (2010) may not generalize to those utilizing the internet to cope with LQTS.

Lieberman and Goldstein (2006) examined the benefits of internet-based list serves for women with breast cancer. Of the 52 women who participated in the study, those who expressed fear and anxiety experienced increased rates of depression and lower quality of life measures, whereas those who expressed anger scored lower on the depression measure and higher on the quality of life measure (Lieberman & Goldstein, 2006). Furthermore, Malik and Culson (2010) reported similar results after surveying 295 women who utilized an online social support group regarding issues with infertility. Over half of the sample reported drawbacks to the online group that included, reading about negative experiences, and reading about other people's pregnancies, wrong

information and also concerns regarding the addictive nature of the online group (Malik & Culson, 2010).

These findings (Liberman & Goldstein, 2006; Malik & Culson, 2010; Salzer et al., 2010) suggest that internet-based peer interactions may be potentially harmful at times. These findings are congruent with the earlier work of Aldwin and Yancura (2004) who theorized that social support is generally positive, unless the respondent reacts negatively, thus creating an increased risk of emotional distress as a result of seeking social support. The benefits of seeking social support as a coping mechanism may be negated if the respondent reacts negatively (Aldwin & Yancura, 2004).

In addition to negative interactions online, other potential drawbacks for use of CMSG have also been reported, specific to those coping with eating disorders. McCormack (2010) conducted a mixed method qualitative and quantitative analysis to better understand the purpose that a CMSG served for people with eating disorders. Three hundred and twenty five messages in 37 discussion topics were included in a deductive thematic analysis. The author reported that the main function of the group was to support others who were coping with an eating disorder. Information seeking was also found to be a common function of the CMSG. Content themes that emerged included: coping with family members, psychological treatment suggestions or recommendations, following a meal plan, and information and personal support for symptoms of the eating disorder. The author cautioned that CMSG can exacerbate an eating disorder if participants are permitted to discuss diet strategies, tips or techniques for purging or restricting. The author recommends that support groups for those coping with eating

disorders should be monitored by a medical professional to help minimize potential negative effects (McCormack, 2010).

The research reviewed here reveals benefits, drawbacks and moderating factors when using CMSG to cope with various medical conditions. Also to be considered is the interpersonal communication style and participant characteristics of internet based group members. The context of seeking support online is different from seeking support in person. Fundamental differences exist when communicating with someone in-person, as compared with communicating with someone via computer mediated technologies, such as the internet. As communications increasingly occur within computer assisted mediums, such as e-mail and instant messages, the effects of these context differences needs to be explored. The participant characteristics and communication styles of those seeking support online for the management of LQTS needs to be considered with the context of computer mediated communication (CMC).

Internet Mediated Communication Styles

Computer mediated communication (CMC) lacks the social context cues that are offered in FtF communications, such as tone of voice, facial expression, indications of socio-economic-status, attractiveness, weight, disability, gender, age, race and sex. It has commonly been thought that this cue deprivation has negative implications for users of CMC. For example, when conducting business via e-mail, the user is forced to make decisions without these context cues that may carry relevant and valuable information. Such socio-cultural context is often necessary to navigate miscommunications and to compensate for potential barriers that may exist. Given these concerns, there is an

increased proclivity for miscommunication when collaborators rely exclusively on CMC for interactions (Cramton & Webber, 2005).

CMC creates a context that contributes to fundamental attribution errors, or a tendency for users to attribute errors because of dispositional factors rather than situational variables (Cramton, 2001). In CMC there is even less information about the situation; attempts to attribute behavior to stable, enduring personality factors are more likely in CMC, as compared with FtF interactions (Gilbert et al., 1998). For example, in an online community that discusses medical information, a participant may be sensitive to cues of intent, judgment or proclivity for support from other users. According to the fundamental attribution error, such things as mistakes in grammar or lack of empathetic statements may be misattributed to character flaws of the user rather than to unobservable situational variables that may exist. In fact, research has indicated that writing style, such as choice of words, grammar, and spelling does impact the impression that CMC users have of each other (Lea & Spears, 1992). Spelling errors were found to influence, negatively, the impression of the sender (Kreiner, Schnakengerg, Green, Costello & McClin, 2002). These findings were further corroborated by Jessmer and Anderson (2001) who found that those who sent grammatically correct e-mail messages were viewed more positively and recipients were found them more desirable to work with.

Benefits of Visual Cue Deprivation.

There are benefits of social and contextual cue deprivation characteristic of CMC. Those from stigmatized racial groups may reap benefits from the visual cue deprivation afforded by CMC. For example, Anderson (2003) found that when CMC is void of visual cues (i.e. not video assisted) the influence of biases related to the ethnicity and race

of the communicator may be reduced. Additionally, Straus, Miles and Levesque (2001) found that less attractive individuals are rated more positively when interactions are conducted without visual cues. These findings suggest that CMC may reduce some biases based on visual and contextual cues, allowing for increased equality and status in computer mediated working relationships.

Gender differences. Medical support groups that discuss psychosocial factors of medical management tend to be composed primarily of women; some studies suggest women outnumber men 4-to-1 in these groups (Krizek, Roberts, Ragan, Ferrara & Lord, 1999). Research also suggests that men and women tend to utilize medical support groups differently; men tend to post and respond to informational content, whereas women will focus and discuss the emotional and psychosocial aspects of their medical concerns (Wolf, 2000). In fact, these findings extend to the way in which men and women consume health related information in general. Research suggests that men may be less likely to seek help from health care providers than women (Addis & Mahalik, 2003; Mackenzie, Gekoski & Knox, 2006) and when they do seek help, they are less likely to report their feelings to health care providers (Corney, 1999). These findings parallel those in the wider literature regarding gender communication differences that suggest men tend to value concrete activities and direct information, whereas women may value emotionally laden conversation and connection with others (Tannen, 1990).

The anonymity afforded of online groups may encourage participants to engage in behaviors that challenge these stereotypical gender norms; for example, some men have displayed an increase in emotional displays online, as compared with their FtF interactions (White & Dorman, 2000). Other research has found that women's discussion

in online medical support communities may adopt a more traditionally masculine role with an increase in assertive and aggressive interaction styles, as compared with their regular FtF interactions (Bellman et al., 1993). Mo, Malik and Coulson (2009) conducted meta-analyses of these gender differences in computer-mediated medical support communities and found that most computer-mediated communication is consistent with gender related norms; however, it is less prominent in mixed-gender support groups. These findings suggest that gender differences are noteworthy in CMC and that the opportunity for anonymity afforded in CMC may encourage some participants to reach outside of their traditional gender-typed communication roles.

Existing online groups for LQTS

Several online sites currently exist for discussion of topics related to LQTS. At least two groups exist on the online social platform; Face book, which is both public and not moderated. Another message board, "QTsydnrome.ch," is available online and is considered public although it is moderated and participants must register to join. Several more internet based user groups for LQTS exist in yahoo groups. These groups range in membership from 1 to 920; they require membership, are public and most have group moderators. The online user group utilized in the current study includes over 900 international members and has existed for about 10 years. Although the group utilized in the current study is public, membership is required. The group is informal, meaning members are free to interact in whatever way they choose. The group includes moderators who are active in topic discussions and review member's posts. The group is marketed as an online support group for those who are affected by LQTS. Topics of

discussion are varied and include but are not limited to genetic testing, new diagnosis and treatment discussions.

Appendix B

Definition of Terms Used

Arrhythmia: a variation in rhythm of the heartbeat either in time or force (Merriam-Webster Medical Dictionary, 2013).

Asynchronous: A type of communication between computers where there is no timing requirement for sending or receiving messages (Merriam-Webster Dictionary, 2011).

Authored Thread: This is a string of posts that originated or was initially authored by an index participant.

Chronic Illness: An illness, disease or syndrome that is recurrent or enduring in duration.

Chronic Medical Condition: Distinguishes chronic illness for chronic medical management and is preferred by some because it explains that those with chronic conditions are not always ill.

Computer Mediated Communication (CMC): Communication that occurs through a computer, such as e-mail, chats or user groups discussion posts.

Computer Mediated Support Group (CMSG)**:** An online user group where members interact and discuss issues of a certain topic areas (such as LQTS).

Face-to-Face (FtF): Communication that occurs in person.

Index Participant: Participant identified for purpose of current study.

Reply Post Contributed: A post offered in reply to a peer authored post.

Sudden Infant Death Syndrome (SIDS): A syndrome where an otherwise healthy infant aged 1 month to 1 year dies suddenly and without explanation (Merriam-Webster Medical Dictionary, 2011).

Synchronous: A type of communication between computers in which a common timing single is established that allows immediate transfer of large bits of data (Merriam-Webster Dictionary, 2011).

Syncope: Loss of consciousness due to inadequate blood flow to the brain (Merriam-Webster Medical Dictionary, 2011).

Appendix C

List of Search Terms Used

New Diagnosis

New Dx

Newly Diagnosed

New Diagnosis

Introduction

New Introduction

New to Group

Intro

New

New Intro

New LQT

New LQTS

New Long QT

My Story

Appendix D

List of Procedural Instructions

Data Collection

1. Data identified through user of search terms (see appendix C).
2. Data downloaded from online user group into a word processing document.
3. Each discussion thread collected as single word document.
4. Each document searched for digital artifacts, such as “carrots” and backslashes.
5. These digital artifacts were electronically removed from each document.
6. Each document searched for redundant posts and these were removed.
7. Each document was electronically searched for participant user names.
8. Identifying information was electronically removed.
9. Data were printed, collated and bound for analysis.
10. Data were double checked and any identifying information.
11. A de-identified copy was prepared for each member of the coding team.

Data Analysis

1. Coding team was formed of three clinical psychology doctoral students.
2. Training in the process of qualitative analysis was provided.
3. Members were provided with a list of coding strategies for reference.
4. Each member was provided with a de-identified copy of the data.
5. Each member conducted an independent coding review.
6. Team meetings occurred every several weeks to review several transcripts.
7. Each member kept process notes to describe the analytic process.
8. Themes were identified at team meetings as each transcript was reviewed.
9. Functional analysis was conducted.

Coding Strategies Used (Corbin & Strauss 2008)

1. Asking Questions
    a. Sensitizing, Theoretical and Practical Questions
2. Making Comparisons
3. Considering Language
    a. Various Meaning of a Word, Indications of Time, Similes and Metaphors
4. Flip-Flopping
5. Drawing Upon Personal Experience
6. Waving the Red Flag
7. Looking for the Negative Case

Appendix E

Rank Order of Emotional Themes

Emotions Expressed in Posts	***Participants who Endorsed Emotion***
Fear, Worry, Uncertainty, Doubt	9
Shock, Numbness, Overwhelmed	3
Frustration, Burdened, Stress	3
Isolation, Discounted, Invalidated	3
Gratitude, Thankful	3
Upset, Concerned (about restrictions)	2
Guilt	2

Appendix F

Rank Order Antecedents to Initial Posting

Event Prior to Posting	*Number of Participants*
Received genetic results	6
Waiting for genetic results	5
Recent cardiac event	3
Preparing for ICD surgery	3
Recovering from ICD surgery	2